well-established.8–10 Pneumothorax is a well-known procedure-related complication. The incidence of pneumothorax has been reported to be between 1% and 5%.9,12 It occurs mostly in relation to transbronchial biopsy procedures.9,12 The standard bronchoscope with an external diameter of ≥ 4.9 mm is not small enough to allow it to reach the peripheral bronchi, so the visceral pleural perforation by the bronchoscope cannot occur.

Recently, ultrathin bronchoscopes have been developed with greater visual range, improved visibility, and a larger working channel. A 2.8-mm diameter ultrathin bronchoscope with a 1.2-mm working channel permits the observation and manipulation of more peripheral bronchi than was previously possible with a standard bronchoscope.1,2 Asano et al2 reported that a 2.8-mm diameter ultrathin bronchoscope (BF-XP40; Olympus) and a 6.3-mm diameter standard bronchoscope (BF200; Olympus) could observe the 5th to 11th generation bronchus (mean [± SD], 7.1 ± 1.5) and the 3rd to 5th generation bronchus (mean, 3.5 ± 0.7), respectively. Thus, the ultrathin bronchoscope showed a significantly greater ability to observe peripheral bronchi.

With the advance of the ultrathin bronchoscope into the peripheral bronchi, however, the visibility of the small airway becomes limited. Therefore, in most cases when an ultrathin bronchoscope is used for performing a biopsy of peripheral pulmonary lesions, it is advanced under fluoroscopic guidance to approach the lesions. This is facilitated by careful tip manipulation of the bronchoscope in the peripheral airway. In our two cases, however, too forceful an advance and manipulation of the bronchoscope under fluoroscopic guidance led to visceral pleural perforation. In ultrathin bronchoscopy, firmly lodging the tip of the bronchoscope into the bronchus, the so-called “wedge technique,” can cause an adverse event. To reduce such injurious complications, manipulations with an ultrathin bronchoscope must be very gently performed, especially in the peripheral lung. Furthermore, to avoid coming too close to the visceral pleura, the position of the tip of the ultrathin bronchoscope should very often be confirmed by fluoroscopy, either by rotating the patient or the arm of a C-arm fluoroscope. The fluoroscopic findings were characteristic. After the ultrathin bronchoscope was passed through the visceral pleura, the tip motion was greater and more unstable than the motion in the peripheral endobronchus.

To our knowledge, the only occurrence of pneumothorax associated with ultrathin bronchoscopy was described in a pediatric patient.11 It was due to transbronchial biopsy using a miniforceps, not the mechanism used in the present report. Thus far, we have conducted 410 transbronchial biopsy procedures using the ultrathin bronchoscope. Pneumothorax has occurred in six patients (1.5%) undergoing all procedures. Two were the present cases, and pneumothorax developed in the remaining four patients following transbronchial biopsy using a miniforceps.

Fortunately, the patients in our cases underwent simple observation and healed spontaneously without the need for invasive treatment such as tube thoracostomy or surgical procedures. Most cases of pneumothoraces related to bronchoscopy have little clinical significance, and life-threatening cases such as those with tension pneumothorax rarely have been reported.11 Needless to say, careful observation is necessary after the occurrence of visceral pleural perforation.

In our two cases, ultrathin bronchoscopy was performed by different bronchoscopists. With the increasing use of ultrathin bronchoscopy, pneumothorax caused by the mechanism described in this report is more likely to occur. To prevent possibly injurious complications, very careful and gentle manipulations of the ultrathin bronchoscope should be performed while confirming the precise position of its tip by fluoroscopy, especially in the peripheral airways.

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Small Cell Carcinoma of the Lung Exclusively Localized Within the Left Descending Pulmonary Artery*

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We encountered a 69-year-old woman displaying a filling defect within the left descending pulmonary artery (PA) on a chest CT scan and pulmonary angiography. A subsequent 2-[18F]fluoro-2-deoxy-D-glucose (FDG) positron emission tomography (PET) scan demonstrated focal uptake in the left hilum. A cytologic examination of transbronchial needle aspiration specimens revealed small cell carcinoma. The patient underwent concurrent radiation therapy and chemotherapy with cisplatin and etoposide, resulting in tumor shrinkage and recanalization of the involved PA. This is the first case of small cell carcinoma localized exclusively within the PA, and positive findings on FDG-PET facilitated the unexpected diagnosis. (CHEST 2005; 127:2273–2276)

Key words: 2-[18F]fluoro-2-deoxy-D-glucose positron emission tomography; intravascular tumor; pulmonary artery; pulmonary thromboembolism; small cell carcinoma; transbronchial needle aspiration

Abbreviations: FDG = 2-[18F]fluoro-2-deoxy-D-glucose; NSE = neuron-specific enolase; PA = pulmonary artery; PET = positron emission tomography; Pro-GRP = pro-gastrin-releasing peptide; PTE = pulmonary thromboembolism; TBNA = transbronchial needle aspiration

Intravascular tumor of the pulmonary artery (PA) represents a rare but important differential diagnosis of pulmonary thromboembolism (PTE). The principal etiology of this clinical entity is primary sarcoma,1,2 with the remaining cases typically representing large tumor embolization from other organs.3 Herein, we describe the first case of small cell carcinoma that was localized exclusively within the PA in which the patient presented with a PA obstruction that is characteristic of PTE. Diagnosis and follow-up were greatly aided by the use of 2-[18F]fluoro-2-deoxy-D-glucose (FDG) positron emission tomography (PET).

CASE REPORT

A 69-year-old woman with uveitis was referred to our department for the investigation of pulmonary sarcoïdosis in December 2002. A chest CT scan revealed no such intrathoracic lesions but detected an unexpected filling defect within the left descending PA (Fig 1, top left, A). No chest symptoms were apparent, but the patient had undergone a panhysterectomy for uterine cancer when she was 45 years old, and a left mastectomy for breast cancer at 66 years old. Laboratory findings showed no clotting disorders. Although conventional chest radiography showed no abnormal shadows, a lung ventilation-perfusion scan demonstrated a perfusion defect in the left lower lung on normal ventilation, and pulmonary angiography revealed complete occlusion of the left descending PA (Fig 1, bottom left, C).

PTE was suspected at this time, but the patient displayed few risk factors for this condition, so an FDG-PET scan was performed to evaluate potential malignancies in other organs. Focal uptake was found only in the left hilum at an identical site to that of the filling defect seen on the CT scan. In addition, a chest CT scan performed 1 month later showed significant enlargement of the lesion, indicating malignant features. At this time, plasma levels of various tumor markers were measured to provide further information regarding the malignancy. Levels of tumor markers were as follows: pro-gastrin-releasing peptide (Pro-GRP), 67.2 pg/mL (normal range, 0 to 10 pg/mL); neuron-specific enolase (NSE), 13.03 ng/mL (normal range, 0 to 10 ng/mL); and carcinoembryonic antigen, 10.3 ng/mL (normal range, 1.0 to 6.5 ng/mL). Bronchial fibroscopy revealed extrinsic compression of the posterior wall of the left lower bronchus. A cytologic examination of transbronchial brush specimens from the lesion surface showed no malignancy, while an examination of transbronchial needle aspiration (TBNA) specimens revealed small tumor cells with scant cytoplasm, finely granular nuclei, and inconspicuous nucleoli, indicating small cell carcinoma (Fig 2, top, A). Cell block sections of tumor cells stained positive for cytokeratin (Fig 2, bottom, B) and thyroid transcription factor-1, while negative results were obtained for vimentin, leukocyte common antigen, synaptophysin, and chromogranin. Since no distant metastatic lesions were identified, the small cell carcinoma of the lung was considered to represent limited disease.

The patient underwent radiation therapy to 45 Gy total with hyperfractionation twice a day at 1.25 Gy, and three concurrent courses of chemotherapy with cisplatin and etoposide, resulting in marked shrinkage of the tumor and recanalization of the involved PA (Fig 1, top right, B, and bottom right, D). By 2 months after undergoing the initial treatment, plasma levels of Pro-GRP, NSE, and carcinoembryonic antigen had normalized to 4.1 pg/mL, 5.40 ng/mL, and 5.4 ng/mL, respectively. No recurrence has been seen as of the time of the writing of this article, >1 year after initiating therapy.

DISCUSSION

We encountered a case of small cell carcinoma that was exclusively localized within the PA. To the best of our knowledge, no similar cases have been reported previously.

We diagnosed small cell carcinoma based on tumor cell size and the nuclear details of TBNA specimens,4 and positive staining for epithelial cell markers, cytokeratin, and thyroid transcription factor-1. Elevated plasma levels of Pro-GRP and NSE supported this diagnosis. The potential influence of crush artifacts had to be considered, but our specimens were sufficiently well preserved to allow the assessment of nuclear status. Although the results of immunohistochemical staining for neuroendocrine markers were negative, this occurs in up to 25% of small cell carcinomas.5

Three possibilities exist with regard to the cellular origin of this small cell carcinoma in the PA. The first is
direct invasion of small cell carcinoma originating from the adjacent bronchus. However, the first chest CT scan showed that the tumor was completely localized within the left descending PA. The second possibility is metastasis or tumor embolus from another organ. Although extrapulmonary small cell carcinoma has been reported on occasion,6 a single metastasis to the pulmonary arterial wall does not appear to represent a natural consequence, considering the high blood flow within the lumen. In addition, no findings on systemic examination, including FDG-PET scanning, indicated the presence of malignancy in other organs. A final possibility is the de novo genesis of small cell carcinoma from vessels of the PA. However, this seems unlikely, as no previous evidence has suggested the existence of neuroendocrine cells in the pulmonary vasculature. Although the cellular origin of this small cell carcinoma in the PA could not be determined, small cell carcinoma exclusively localized within the PA is very rare, and we hope that other similar reports will pursue and clarify the origins of such tumor in the future.

In the present case, FDG-PET scanning provided crucial information for early diagnosis, since the second CT scan and TBNA were performed based on abnormal findings from the FDG-PET scan. Delayed diagnosis would complicate the clarification of the relationship between the tumor and the PA. Small cell carcinoma exclusively localized within the PA might thus have been overlooked in the past.

**Conclusion**

The present case indicates that small cell carcinoma can be localized within the PA, with the patient presenting with PA obstruction as seen in patients with PTE. FDG-PET scanning proved extremely useful for clarifying this unexpected diagnosis.
Silicone Embolism Syndrome

A Case Report, Review of the Literature, and Comparison With Fat Embolism Syndrome*

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Liquid silicone is an inert material that is utilized for cosmetic procedures by physicians as well as illegally by nonmedical personnel. We present a case report and collated clinical findings of 32 other patients who were hospitalized after illegal silicone injections. Symptoms and signs of the “silicone syndrome” included dyspnea, fever, cough, hemoptysis, chest pain, hypoxia, alveolar hemorrhage, and altered consciousness. Bilateral patchy alveolar infiltrates were present on the chest radiographs, and silicone pulmonary emboli were detected in all the patients. The patients could be divided into two groups based on the initial presentation and clinical outcome. Twenty-seven patients in group 1 presented predominantly with respiratory symptoms, and 93% of patients were discharged home within 3 weeks. Six patients (group 2) presented with severe neurologic findings, and experienced rapid deterioration and 100% mortality. The clinical findings after silicone embolism are very similar to the published reports of fat embolism, including hypoxemia in 92% of patients with silicone embolism (patients with fat embolism, 56 to 96%), dyspnea in 88% of patients (patients with fat embolism, 56 to 75%), fever in 70% of patients (patients with fat embolism, 23 to 67%), alveolar hemorrhage in 64% of patients (patients with fat embolism, 66%), neurologic symptoms in 33% of patients (patients with fat embolism, 22 to 86%), petechiae in 18% of patients (patients with fat embolism, 20 to 50%), chest pain in 15% of patients (patients with fat embolism, 20 to 50%), and mortality in 24% of patients (patients with fat embolism, 5 to 20%). The similarities among the mode of injury to the lung, the clinical findings, and the high incidence of alveolar hemorrhage suggest a common pathogenesis of silicone and fat embolism syndromes. We discuss the possibility that the activation of the coagulation system may be important in the development of these clinical syndromes.

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Figure 2. Top, A: cytologic examination of TBNA specimens. Numerous small tumor cells with increased nucleocytoplasmic ratio and high chromatin density are apparent (Papanicolaou, original \( \times 800 \)). Bottom, B: histologic examination of cell-block sections from TBNA specimens. Cytoplasm of atypical cells is stained brown (anti-cytokeratin antibody, original \( \times 800 \)).